

Recurrence of Adenoid Cystic Carcinoma in the Breast After Lumpectomy and Adjuvant Therapy

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Adenoid cystic carcinoma (ACC) of the breast is very rare, accounting for only 0.1% of all breast carcinomas.¹⁻⁴ This unusual neoplasm of the breast has certain histopathologic and biological characteristics that distinguish it from more common histologic types. For example, the prognosis of ACC appears to be very favorable. The incidence of axillary lymph node involvement is lower, and distant metastases are uncommon, although they can occur without previous lymph node involvement. Clinical experience has shown that this entity has a low propensity for local recurrence after excision.^{4,5} We present a case of local recurrence of ACC in the breast detected by ultrasonography after successful treatment by lumpectomy followed by adjuvant chemotherapy and radiotherapy.

Abbreviations

ACC, adenoid cystic carcinoma

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Case Report

A 48-year-old woman came to our hospital because her screening mammography showed dense mammary tissue with a poorly defined mass in the upper center of the right breast. Adjunctive breast ultrasonography performed at our hospital showed a mass indicative of malignancy in the same location (Figure 1). An ultrasonographically guided 14-gauge core needle biopsy was taken, and pathologic diagnosis confirmed ACC. The patient underwent lumpectomy of the right breast and dissection of right axillary lymph nodes. The specimen of breast measured $2.7 \times 5 \times 3.4$ cm and contained a poorly defined, pinkish tan, round mass measuring 1.3×1.2 cm. The surgical margins were free of tumor, and all 14 dissected lymph nodes were negative for tumor. The final pathologic report was ACC, and the estrogen receptor, progesterone receptor, and c-erb-B2 status of the tumor were all negative. The patient received adjuvant chemotherapy with a 6-cycle regimen of cyclophosphamide-methotrexate-5-fluorouracil and radiotherapy to the right breast in a daily fraction of 1.8 Gy for a total of

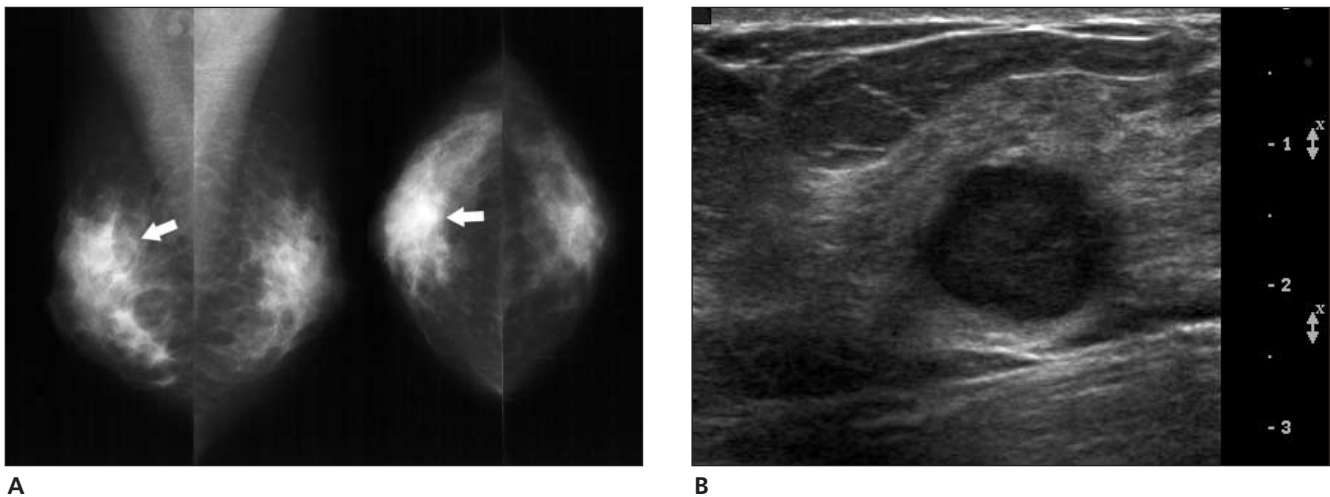


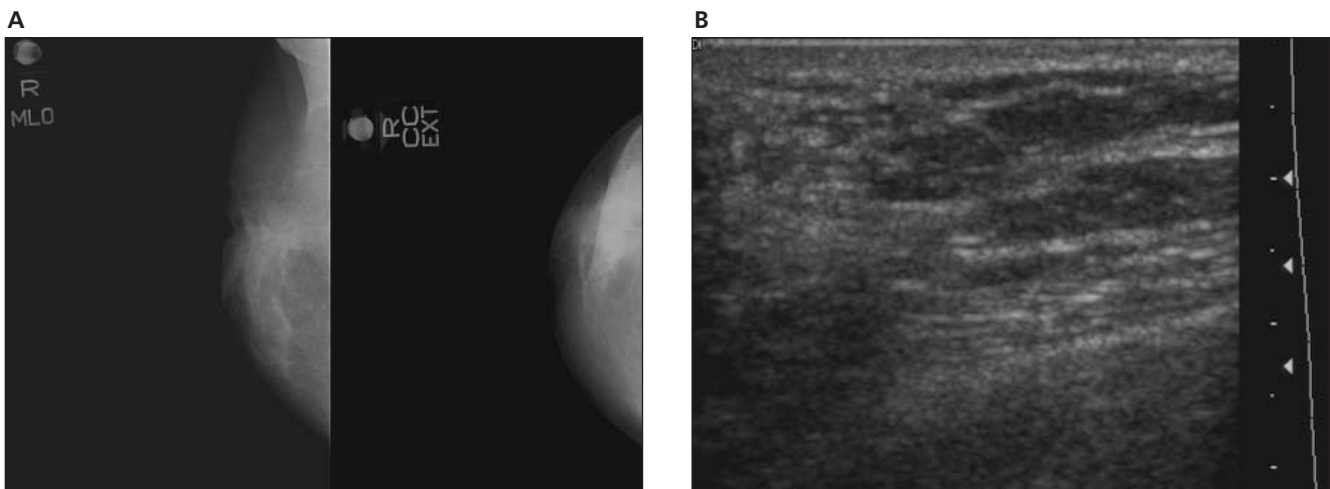
Figure 1. Mammograms and sonogram obtained 18 months before recurrence. **A**, Mediolateral oblique (left) and craniocaudal (right) mammograms show a poorly defined, round, hyperdense mass in the upper center of the right breast (arrows). No abnormalities are seen in the left breast. **B**, Sonogram reveals a 1.3-cm, round, microlobulated, hypoechoic mass with posterior acoustic enhancement in the 12-o'clock location.

59.4 Gy (50.4 Gy to the whole breast and then a 9-Gy boost to the tumor bed area). The patient tolerated the treatment well and had no evidence of recurrence on the follow-up mammography or ultrasonography until 6 months after the adjuvant therapies.

The patient had localized tenderness in the periareolar aspect of the right breast 1 year after lumpectomy of the ipsilateral breast. Physical examination found only localized tenderness without any evidence of a focal mass at the tender site. Follow-up mammography and targeted

ultrasonography of the tender area showed no abnormalities (Figure 2). After 6 months, there was aggravated tenderness at the lumpectomy site, and ultrasonographic examination showed a 6-mm, round, microlobulated, hypoechoic nodule at the lumpectomy site. The tenderness was aggravated as the probe compressed the nodule (Figure 3). The patient underwent an ultrasonographically guided 14-gauge core needle biopsy, and ACC was diagnosed again. A simple mastectomy was performed, and the recurrence of ACC was confirmed pathologically.

Figure 2. Follow-up mammograms and sonogram obtained 1 year after lumpectomy and adjuvant therapy. **A**, Right mediolateral oblique (left) and craniocaudal (right) mammograms show postoperative changes of the breast deformity, distortion, and increased densities due to fibrotic scar formation. Therefore, it is difficult to differentiate them from any possible recurring lesion. **B**, Sonogram of the area of localized tenderness. The upper medial portion of the right breast depicts no abnormality.



Discussion

As with all types of breast cancer, treatment options have evolved for patients with ACC of the breast and depend on the circumstances of a given patient. Wide excision alone could be curative in most cases because of the favorable biological behavior of this disease.⁶ The case presented here is notable as a local recurrence of ACC in the breast even after successful treatment with lumpectomy, adjuvant chemotherapy, and radiotherapy. Few articles have described adjuvant radiotherapy after initial local excision, and, to our knowledge, there has been only 1 case of local recurrence of ACC in the breast after lumpectomy followed by adjuvant radiotherapy, suggesting that radiation after lumpectomy may play a complementary role for local control.^{4,5,7} Little mention is made in the literature of adjuvant systemic therapy for this diagnosis, and clear indications for the role of adjuvant systemic therapy remain to be defined.

Because of the rarity of this entity, there is little information about the characteristic features of ACC on mammography or ultrasonography. Santamaria et al⁸ reported that on mammography this tumor often appears as moderately circumscribed, lobulated nodules that are similar to other types of benign and malignant tumors. In 2 cases of ACC described by Sheen-Chen et al,⁹ ultrasonography showed nodules with well-defined margins and heterogeneous echogenicity without the characteristic changes of an infiltrating carcinoma. Therefore, a benign-looking breast lesion in radiologic examinations cannot completely exclude the existence of this disease⁹; some tumors cannot be identified on mammography because of increased density of the breast.^{8,9} Ultrasonography can identify and help characterize a lesion in the breast with a predominance of dense fibroglandular tissue that could be missed on mammography.¹⁰

In this case, the first targeted ultrasonography for localized tenderness failed to depict a recurring lesion. According to Leung et al,¹¹ most of the ultrasonographic examinations performed to evaluate focal breast pain yielded negative findings, and no cancer was identified. They concluded that the inappropriate use of targeted ultrasonography for focal breast pain should be a reason for concern in the absence of evidence-based data that would justify its use. In a study by McClenathan and de la Roza,¹² however, pain was

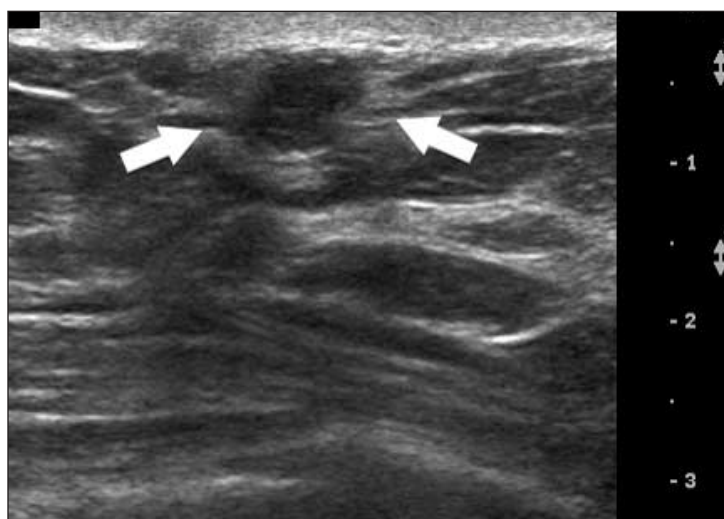


Figure 3. Sonogram obtained 6 months after image in Figure 2 shows a new palpable nodule just at the tender site of the right breast (arrows).

a prominent symptom of patients with ACC, several of whom were monitored for localized pain in the breast for more than 1 year before a mass or radiographic abnormality was detected. Therefore, if localized tenderness or pain occurs with or without a mass, ACC of the breast should be included in a differential diagnosis. In our case, localized pain was the first sign of recurrence of ACC without previous symptoms of localized pain or tenderness related to the primary lesion. Despite little information about the recurrence of ACC, localized pain could be a new symptom, regardless of previous symptoms. If there is a painful sensation as the probe compresses the lesion on follow-up ultrasonography after treatment of ACC, this unusual sign could be indicative of its recurrence, whether there was localized pain related to primary ACC. Suppose additional magnetic resonance imaging was performed when localized pain presented but targeted ultrasonography did not show a recurring lesion; in such a case, it might be helpful for detection of a recurring lesion. Until now, to our knowledge, the findings of ACC in the breast on magnetic resonance imaging have not been reported, and performance of magnetic resonance imaging has been uncertain for the detection of recurrence of ACC. Further study regarding this is needed. Local recurrence, although rare and generally occurring long after the initial event, should be considered in a patient with a history of ACC in the breast, and appropriate follow-up and clinical correlations could be helpful for early detection of a possible recurrence.

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